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Simultaneous Occurrence of Intracranial Aneurysm and Arteriovenous Malformation

by

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Both intracranial aneurysm and arteriovenous malformation which may have a common congenital basis occur seldom simultaneously.

We have recently cared for such a patient. He was found to have an aneurysm of the left anterior cerebral artery, and an arteriovenous malformation of the left fronto-parietal area fed by the same artery. Surgical treatment was successfully performed without bewailed residual neurological signs except for hypo-osmia.

Because of the paucity of such cases in the literature, this case is being reported.

CASE REPORT

K. H., a 36 year-old factory worker was admitted to our Clinic on January 18, 1967. About three months prior to admission, there developed severe headache and vomiting in defecation followed by unconsciousness for a week. He was conservatively managed as subarachnoid hemorrhage in another hospital. After the complete recovery from that episode, no relapse of the attack had taken place until he was referred to our Clinic. As there had been more than three months after the attack of subarachnoid bleeding at the time of admission, remarkable neurological symptoms were absent except for intermittent heavy sensation in his head, especially in occipital and nuchal region.

So far as he informed, he had been in good health until above-mentioned episode.

The patient was a well developed, righ-handed man. Neither nuchal rigidity nor weakness was demonstrated at the time of admission. The tendon reflexes were slightly hyperactive, but no pathological reflexes were elicited. Blood pressre was 112/60 mmHg. On lumbar puncture the initial pressure showed 210 mm H_2O in unilateral position, and protein in cerebrospinal fluid was found to be slightly increased (67 mg/dl) without xanthochromia.

Skull and chest roentgenograms were normal. Electroencephalogram disclosed slight slowing in all leads, while bilateral carotid angiograms revealed an angioma of the left frontoparietal area, fed by the dilated anterior cerebral artery. In addition, a saccular aneurysm was noted on the same artery at the junction of the callosomarginal artery

(Fig. 1 and 2). These lesions were also demonstrated by contralateral angiogram (Fig. 3). For the prevention of rupture of aneurysm or angioma, the operative procedure was planned to attempt the ligation of the feeding artery.

Operation : On February 6, 1967, the bifrontal craniotomy was done under general hypothermia, with use of intermittent positive-negative hyperventilation and continuous spinal drainage. A bilateral horse shoeshaped coronal scalp incision was put, and scalp and the bone flap were reflected down with its base forward the face. After the incision of dura, the site of the first portion of the left anterior cerebral artery become enough visualized. The two silver clips were placed on the most proximal part of the second portion of the left anterior cerebral artery, just before it entered the interhemispherical fissure. On the approach to this region the bilateral olfactory nerves were damaged.

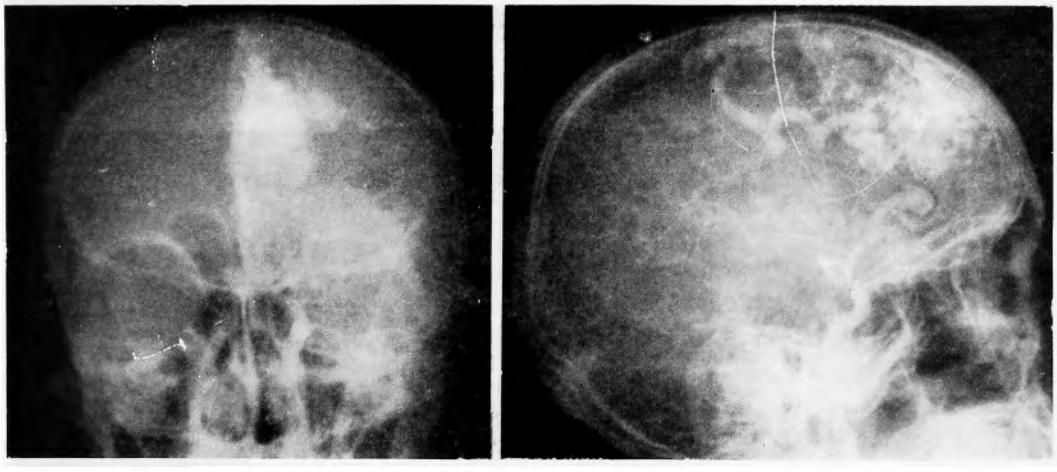

Fig. 1 Left carotid angiograms
Anteroposterior and lateral views showing a saccular aneurysm at the knee of pericallosal artery and angiomatous malformation at its peripheral area

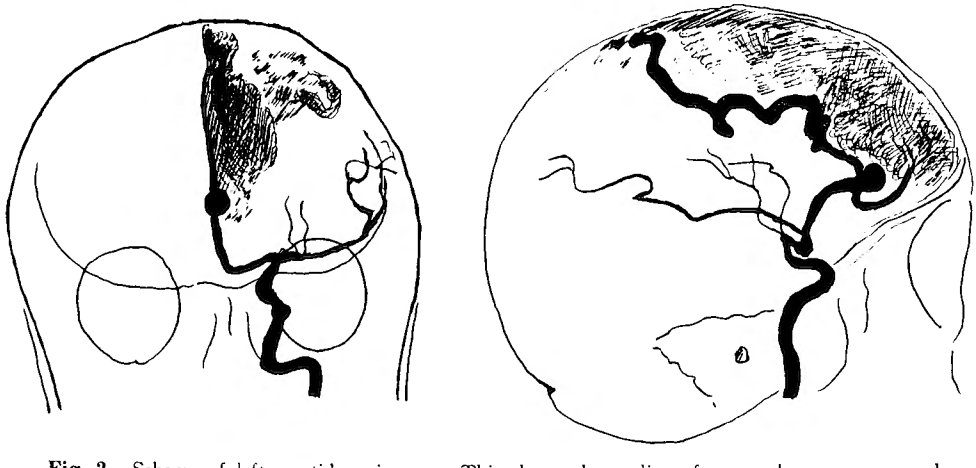

Fig. 2 Schema of left carotid angiograms. This shows the outline of a saccular aneurysm and angiomatous malformation.

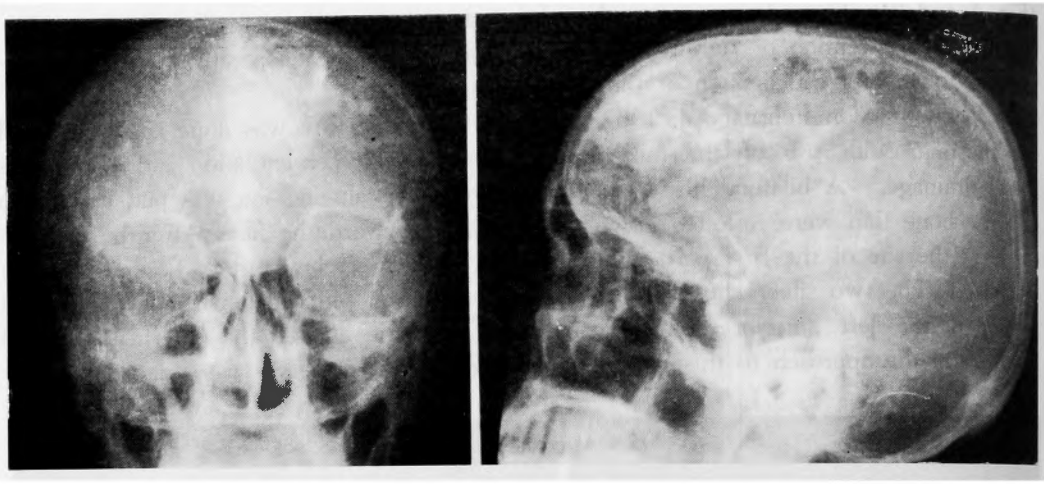

Fig. 3 Right carotid angiograms.

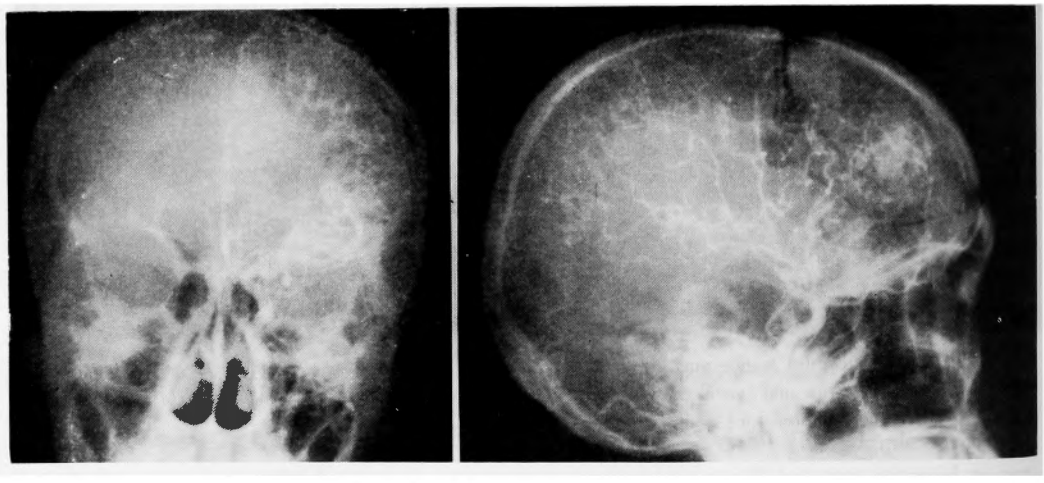

Fig. 4 Postoperative carotid angiograms
The two silver clips interrupt the blood flow of the anterior cerebral artery except for A_1 portion. The aneurysm disappears completely.

The patient made a rapid recovery and was out of bed at the seventh day after the operation. There were no obvious mental or neurological symtoms except for hypo-osmia which was able to be verified only by objective examination.

Nineteen days after the operation when the patient was practically free from symptoms, left carotid angiography was repeated in order to determine the effect of the ligation of the left anterior cerebral artery. The aneurysm and the hypervascular malformation were no longer visualized in the lateral and anteroposterior views (Fig. 4).

However, the late carotid angiogram which was carried out three months after the operation revealed the faint picture of the angiomatous malformation though the aneurysm disappeared completely. The reappearance of the angioma is probably due to the formation of collateral anastomosis while the risk of rupture is considered to be greatly diminished

because of the interruption of blood supply into the aneurysm. The patient is in good health and free from mental or neurological signs except for hypo-osmia.

DISCUSSION

Simultaneous occurrence of aneurysm and arteriovenous malformation is a rarity. According to the other cases in literature the incidence of coexistence is taken to be about 1 to 4 per cent[11)12)]. It is assumed that these lesions originate as a congenital maldevelopment of the blood vessels in early embryonic existence.

Intracranial aneurysms are more frequently found in the third decade. The site of localization in most case is on supratentorial region especially anterior communicating artery and middle cerebral artery, but it is reported that when locating on anterior cerebral artery the aneurysm exists mostly in the peripheral portion from a turnig point of anterior communicating artery[14)].

On the other hand, the arteriovenous malformation has a higher incidence in younger decade than in aneurysms. Localization of arteriovenous malformation is in the overwhelming majority on middle cerebral arterial area. If including the arteries concerned with the lesion, its frequency occurring in middle cerebral arterial area is estimated approximately 70 per cent[15)].

Comparing with those coexistent cases reported by others[1)2)3)4)9)10)11)] our present case also has a single saccular aneurysm on the artery feeding the angioma. In 1956, PATERSON and MCKISSOCK[11)] suggested that the increased blood flow in consequence of the presence of the arteriovenous malformation might contribute to the formation of the aneurysm by giving the abnormal stresses on the cerebral vessels.

However, there is an objection to this hypothesis on the basis of difficulty in explaining the case which have both aneurysm and angioma in separate territory of the different cerebral arteries[7)10)]. It is reported in some literatures[1)12)17)] that one-third or nearly half of the aneurysms were on arteries not feeding the angioma. Being deduced from the variety of the aneurysmal forms it might be more reasonable to consider the generally mentioned etiologic factors, i. e. congenital abnormalities of the vascular muscularis, diffuse (or localized) arteriosclerotic changes, bacterial and/or other inflammations.

Surgical treatment should be required in the patients suffering from recurrent hemorrhage, convulsive seizures and so on. But it is likely that the size and the site of the lesion determine the operability with the condition of the patients.

For aneurysm, the clipping of the neck, trapping, ligation or plastic coating are generally performed according to its form and site. Recently the artificial thrombosis is made an attempt physically or bioelectrically getting a hint from rheology [5)8)].

For arteriovenous malformation, the total extirpation is the choice of surgery. However, as the total extirpation is often impossible when the lesions exist in deep regions of basal ganglia and internal capsule, or those lesions widespread when removing the angioma which are on the motor area of the major hemisphere, it is the problem whether the permanent neurological deficit inconvenient for the social life would occur.

According to POOL[13)] the applicability of the total extirpation is assumed to be about 50 per cent. Furthermore SVIEN and MCRAE[16)] reported that the indication for operative

treatment is restricted in less than 20 per cent.

Upon these facts, for the patients who are difficult to excise, the artificial embolization is well known to use in some cases conservatively. Meanwhile, there are also the opinions[6] that, even if the lesion exists in the important cortical region except for in the impossibly deep region, the radical extirpation is necessary because of the prevention for recurrent hemorrhage and/or the potentially existing ischemia. In any case, if the lesion is located in a suitable area, the radical extirpation should be undertaken. Although the possibility of the occurrence of the brain infarction after ligation or clipping would remain, the ligation of the artery feeding the arteriovenous malformation should also be taken into consideration, for the patients whose lesion is unable to remove completely, as a second best stratagem. It seems that the postoperative brain infarction, however, may not occur as the postoperative blood supply will be compensated collaterally from arteries of opposite hemisphere or branches of the external carotid artery.

As above mentioned, in this case the clipping for two lesions was preferred to hazardous techniques since the arteriovenous malformation is too extensive to excise completely.

SUMMARY

1. An unusual case is roported of a 36 year-old man with the coexistence of intracranial aneurysm and arteriovenous malformation.

2. It is assumed that both forms of vascular lesions arise from the congenital maldevelopment of the blood vessels in early embryonic existence.

3. Some problems concerned with surgical treatment are discussed.

4. In the present case more than nine months after operative intervention, he is in good health and working at factory.

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和文抄録

脳動脈瘤及び動静脈奇形の共存せる1例

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入院3ヵ月前にくも膜下出血の発作があり，ひきつづいて約1週間の意識障害のあつた36才の男子に，左前大脳動脈瘤及びこの動脈を栄養血管とするかなり広範囲にわたる動静脈奇形を認めた．動静脈奇形に対しては全剔出することが理想ではあるが，これが困難と思われたので次善の策として動脈瘤を含んだ中枢側でclipping を試みた，手術に際して両側の嗅神経を損傷せしめたことはさけられなかつた．

術後は hypo-osmia を示した外には何ら神経学的欠損症を残すことなく経過し，術後19日目の脳血管写では，ほぼ動静脈奇形の像は消失し，動脈瘤は完全に消失していた．いずれ動静脈奇形の方は副血行路の新生で再発して来る可能性は充分考えられるが，動脈瘤からの出血による死亡率が動静脈奇形からのそれよりもはるかに高いことから考えればclipping の効果は大いに期待出来ると考えられる．

現在，術後9ヵ月になるが何ら日常生活に支障をきたさず工場に勤務している．

以上比較的稀と思われる脳動脈瘤及び動静脈奇形の合併例を報告した．